

Aorto-iliac aneurysm associated with congenital pelvic kidney: A short series of successful open repairs under hypothermic selective renal perfusion

Enrico Maria Marone, MD, Yamume Tshomba, MD, Chiara Brioschi, MD, Fabio Massimo Calliari, MD, and Roberto Chiesa, MD, *Milan, Italy*

The occurrence of congenital pelvic kidney (cPK) during aorto-iliac aneurysm repair is an extremely unusual finding. We report a series of four patients with aorto-iliac aneurysm and associated cPK who underwent aorto-iliac repair at our institution over the last 10 years. Aorto-iliac aneurysm repair under cPK selective hypothermic perfusion was successfully accomplished in all cases. All the cPK arteries were spared and were selectively reimplanted when required. No major complications or death were reported at long-term follow-up. Open surgical repair of aorto-iliac aneurysm in patients with cPK is safe and effective and, in our short series, we observed no worsening of the renal function; besides, we reported a persistent improvement of the renal function in two out of the four cases. (*J Vasc Surg* 2008;47:638-44.)

The occurrence of a pelvic kidney is an uncommon condition with an incidence between one in 2100 and one in 3000 births.¹ The association of pelvic kidney with aorto-iliac aneurysm disease is an extremely rare clinical setting and only 17 cases of aorto-iliac open repair have been previously reported²⁻¹⁰ (Table I) with a variety of techniques and adjuncts of renal protection. We present our single center experience of successful open repair of aorto-iliac aneurysms with associated cPK.

MATERIALS AND METHODS

From January 1993 to January 2007, at the Scientific Institute San Raffaele of Milan, 3223 patients underwent elective aorto-iliac aneurysm open repair. Among these large series, in four cases a congenital pelvic kidney was associated. The main features of these four patients are summarized in Table II.

In this short series aneurysm and renal arteries, specific assessment was preoperatively performed in all cases by means of computed tomography or magnetic resonance associated with renal arteries angiography and duplex ultrasonography. Perioperative renal function changes were monitored by creatinine serum level assessment. In three hypertensive patients, base-line and captopril radionuclide preoperative and postoperative renographic study were also performed.

In all cases, the intervention was performed through an elective midline transperitoneal approach. The aorta was always cross-clamped under systemic heparinization (70 IU/Kg). Before and during cross-clamping, the diuresis was forced in all cases by systemic infusion of furosemide

and mannitol 18% (12.5 g). The aneurysms were incised longitudinally, thrombus was removed, and the orifices of the cPK arteries were visualized. During aortic repair, for the duration of the cross-clamping, a bolus infusion of 200 ml of cold crystalloid solution (lactate Ringer 4°C + mannitol 18% 70 ml, 6-methylprednisolone 500 mg in 500 ml) was infused into the orifices of the cPK arteries through an occlusion-perfusion balloon catheter (Pruitt-Inahara, LeMaitre Vascular, St. Petersburg, Fla) by means of infusible pressure infusor,^{11,12} then continuous perfusion was infused in attempt to reduce the temperature of the kidney to 15° to 18°C (Fig 1). Aneurysm repair was accomplished in all cases by means of endoaneurysmal technique with Dacron tube or bifurcated grafts.

RESULTS

Preoperative image studies showed normal parenchyma in all the cPK except for a patient (case 4) that presented a cPK cystic dysplasia with cysts with diameter variable from 0.5 to 14 cm with a small amount of functioning parenchyma. No significant cPK renal arteries stenosis or kinking or significant renal veins and urinary excretory system compression or other anomalies were preoperatively detected. Aneurysm repair was successfully accomplished in all patients.

In case 1, a right cPK presented a single feeding artery originating from the distal aorta immediately above the aortic carrefour. Proximal aortic cross-clamping was performed below the single contralateral renal artery, and the aortic repair was performed with a Dacron tube graft. The selective hypothermic perfusion into the cPK artery ostium was carried out until the completion of both proximal and distal anastomosis in which the cPK artery ostium was included (Fig 1).

In case 2, a left cPK presented a main artery originating from distal aorta and an inferior polar artery originating from right hypogastric iliac artery providing arterial circulation also to the inferior right kidney. The aortic cross-clamping was performed below the upper right renal artery

From the Department and Chair of Vascular Surgery, San Raffaele Scientific Institute, Università Vita-Salute.

Competition of interest: none

Correspondence: Enrico Maria Marone, MD, Chirurgia Vascolare, IRCCS San Raffaele Via Olgettina, 60, Milano, 20132, Italy (e-mail: e.marone@hsr.it).

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Table I. Table showing the literature review of patients underwent aorto-iliac aneurysm repair with associated congenital pelvic kidney

<i>Author</i>	<i>No. of cases</i>	<i>Side</i>	<i>Technique of repair</i>	<i>Renal preservation</i>
Ezzet et al ³⁰	1	Left	Dacron bifurcated graft (pelvic kidney artery originating from right common iliac artery)	Simple clamping
Hans and Robb ²	1	Right (right renal vein from the pelvic kidney joined the right common iliac vein)	Dacron bifurcated graft	In situ hypothermic flushing with heparinized ice-cold saline (2 feeding vessels arising from both the common iliac arteries being left intact distally to the iliac anastomoses)
Hollis Jr et al ³	2	1 right	Dacron bifurcated graft (aortoiliac aneurysm); 2 of 2 arteries reimplanted ("Carrel patch") to the main body of the Dacron graft	Proximal double clamping during proximal anastomosis and in situ hypothermic perfusion during distal anastomosis
		1 left	Dacron tube (AAA) graft, the lower of 2 arteries included in the distal aortic anastomosis and the upper reimplanted in the left common iliac artery	Selective hypothermic perfusion
Schneider et al ⁴	1	Left	Dacron bifurcated graft (aortobiliac aneurysm); 1 of 1 artery reimplanted to the right iliac limb of the Dacron graft	Proximal double clamping during proximal aortic anastomosis and passive shunt (Javid) from the Dacron graft to the pelvic kidney artery during distal iliac anastomoses and renal artery reimplantation
Glock et al ¹³	1	Right	Dacron tube graft (AAA), 1 of 2 arteries reimplanted (saphenous vein graft), the other was included in the distal aortic anastomosis	Double distal clamp
Mandolfino et al ⁵	1	Right	Dacron bifurcated with 2 renal arteries reimplanted to the graft	Simple clamping with systemic infusion of dopamine and mannitol
Murakami et al ⁶	1	Left (associated malrotation of the gut and left undescending testis)	Dacron tube graft (2 pelvic kidney arteries included in distal anastomosis)	Hypothermic selective perfusion of the upper renal artery via an 8 French size Foley balloon catheter + ice slush applied over the kidney (orifices of renal arteries left intact to the aortic wall)
Hanif et al ⁷	1	Right	Dacron trifurcated graft for aortoiliac aneurysm repair and selective grafting of 1 of 1 ectopic artery (9 mm Dacron graft)	Passive shunt (10 mm Dacron graft with a Javid-IMPRA Inc., USA- secured to its distal end) from the right axillary artery to the pelvic kidney artery during aortic anastomosis, iliac anastomoses and renal artery selective grafting
Renzulli et al ⁸	1	(horseshoe)	Dacron tube graft with superior mesenteric artery selective bypass graft (TAAA)	Distal double clamping during proximal aortic anastomosis and hypothermic selective perfusion of the largest renal artery during distal aortic anastomosis + ice slush applied over the kidney
Faggioli et al ³¹	3	NA	Ectopic renal arteries reimplantation or inclusion in the distal anastomosis	Selective renal hypothermic perfusion for cross-clamp time >40 min, no renal protection systems for cross-clamp time < 40 min
Meyer et al ¹⁰	2	1 left, 1 right	NA	NA
Krohn et al ³²	1	(horseshoe)	Staged bilateral iliac Dacron graft (bilateral iliac artery aneurysm with multiple feeding vessels arising proximally to the aneurysm and from the hypogastric arteries)	Staged bilateral selective common iliac → hypogastric artery bypass graft to reduce the hypogastric clamping time (bilaterally < 30 min) followed by selective revascularization of external iliac arteries

Table I. *Continued.*

<i>Author</i>	<i>No. of cases</i>	<i>Side</i>	<i>Technique of repair</i>	<i>Renal preservation</i>
Rehrig Jr et al ⁹	1	Right	Dacron bifurcated graft (bilateral iliac artery aneurysm)	Proximal double clamping during proximal aortic anastomosis and passive shunt (Baxter Healthcare Corp, Waukegan, IL) from the main body of the Dacron graft and the pelvic kidney artery during iliac anastomoses and renal artery selective reimplantation
Marone et al ¹⁴	4	2 right, 2 left (1 functioning multicystic kidney)	2 Dacron tube graft (AAA) 2 Dacron bifurcated graft (1 aortoiliac aneurysm and 1 right iliac aneurysm), 2 renal arteries reimplanted, 4 included in the proximal or distal anastomoses	Hypothermic selective perfusion (Pruitt Irrigation Occlusion catheter 9 Fr.-LeMaitre Vascular)
Total	21			

AAA, Abdominal aortic aneurysm; NA, not available.

Table II. The table shows the main features of our series of four cases of a congenital pelvic kidney

<i>Case</i>	<i>Age</i>	<i>Sex</i>	<i>Aneurysm site (diameter mm)</i>	<i>Pelvic kidney side (functioning)</i>	<i>Feeding arteries</i>	<i>Mean preoperative blood systolic/diastolic pressure (blood pressure therapy)</i>	<i>Blood systolic/diastolic pressure at late follow-up (blood pressure therapy)</i>
1	72	F	Infrarenal aorta (55)	Right (perfusion impairment)	Single originating from right common iliac artery	170/100 mm Hg (losartan 50 mg/day)	Normotensive (none)
2	68	M	Infrarenal aorta (54)	Left (perfusion impairment)	Main artery originating from distal aorta and inferior polar artery originating from right hypogastric iliac artery	170/100 mm Hg (nifedipine 120 mg/day)	Normotensive (none)
3	63	F	Right common iliac artery (39)	Right (well functioning)	Single originating from abdominal aorta	120/70 mm Hg (none)	Normotensive (none)
4	61	M	Infrarenal aorta (58) and left common iliac artery (39)	Left (perfusion impairment and multicystic dysplasia)	Main artery originating from distal aorta and inferior polar artery originating from left common iliac artery	150/90 mm Hg (atenolol 25 mg/day and hydrochlorothiazide 25 mg/day)	140/85 mm Hg (atenolol 50 mg/day and hydrochlorothiazide 25 mg/day)

and the aortic repair was performed with a Dacron tube graft. A selective hypothermic perfusion of the upper cPK artery was carried out until its reimplantation to the tube graft after the completion of the proximal aortic anastomoses (Fig 2).

In case 3, a right common iliac artery was treated. In this case, the associated cPK was perfused by a single feeding artery originating from distal aorta. The aortic cross-clamping was performed below the single contralateral renal artery and above the cPK feeding artery.

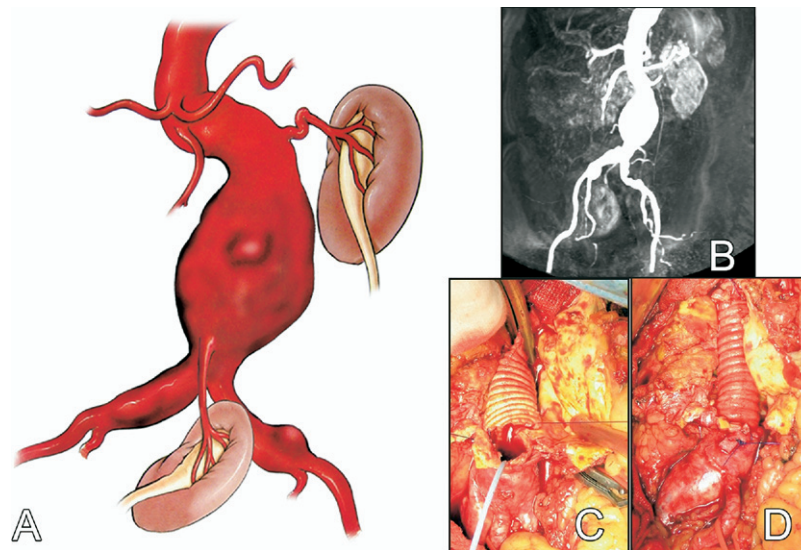


Fig 1. A case of infrarenal aortic aneurysm associated with a cPK with a main artery originating from distal aorta and an inferior polar artery originating from contralateral hypogastric artery providing arterial circulation also to the contralateral kidney. Preoperative 3D computed tomography scan (A) and schematic illustration (B). Also the main stages of operation are illustrated: perfusion of cold crystalloid into the orifices of the cPK main artery through an occlusion perfusion balloon catheter (C), and end-to-end Dacron aortic bypass with the main cPK artery reimplantation (D).

For the aneurysm repair, a Dacron bifurcated graft was used. The hypothermic perfusion of the cPK artery was carried out until the completion of proximal anastomosis, allowing the iliac anastomosis to be performed under direct hematic perfusion of the main cPK artery.

In case 4, a polycystic cPK presented a main artery originating from distal aorta near the aortic bifurcation and, additionally, an inferior polar artery originating from distal left common iliac artery. The aortic cross-clamping was performed below the single contralateral renal artery. Reconstruction required the use of a Dacron bifurcated grafts. After larger cysts drainage and excision to allow the surgical access, the hypothermic perfusion of the main cPK artery was carried out until its reimplantation to the main body of the bifurcated graft after the completion of the proximal aortic anastomoses and the iliac anastomoses were performed under direct hematic perfusion of the main cPK artery. The distal end-to-end anastomosis was first performed with the left common iliac artery, above the orifice of the inferior polar artery, and later with the right common iliac artery. During aortic/left iliac anastomosis and main cPK artery reimplantation the inferior cPK polar artery was perfused by backflow.

The overall mean operative time was 178 minutes (range 133 to 251 minutes), the overall mean aortic cross-clamping time was 32.4 minutes (range 22 to 53 minutes), and the overall mean duration of cPK cold ischemia was 15.3 ± 4.9 minutes (range 8 to 19 minutes). After the bolus infusion of 200 ml, a mean of 230 ± 136.1 ml (range 180 to 420 ml) of lactate Ringer at 4°C was perfused into the orifices of the cPK arteries. In all the cases, intraopera-

tive auto-transfusion of blood was used. Use of packaged concentrated red blood cell transfusion was required in two cases with a mean of 1.5 units for patient (range 1 to 2 units).

No patient had permanent or transient renal function impairment. Postoperative courses were uneventful in three cases. Patient 1, with a past medical history of arterial hypertension, paroxysmal atrial fibrillation and previous coronary angioplasty, presented a single episode of atrial fibrillation successfully treated with medical therapy. Renal arteries color-flow Duplex at discharge showed in all the cases a successful aneurysm repair and patent cPK feeding arteries with good perfusion of the cPK cortex.

Preoperative base-line and captopril radionuclide renographic studies performed in the three hypertensive patients revealed the renovascular nature of the hypertension in all the cases. Two preoperatively hypertensive patients had postoperative captopril-renogram and serum creatinine levels returned to normal and were discharged normotensive without blood pressure medication. The normotensive patient with preoperative normal renal function experienced an initial improvement of serum creatinine levels, and a return to the base-line values at the discharge. The patient with the polycystic cPK did not experience significant serum creatinine levels and captopril-renogram improvement following aorto-iliac repair and was discharged under the same blood pressure.

At a mean follow-up time of 43 ± 77.4 months (range 6 to 168), we did not register loss of renal artery branches, neither segmental renal infarcts at computed tomographic scan. No evidence of new onset hypertension or changes in

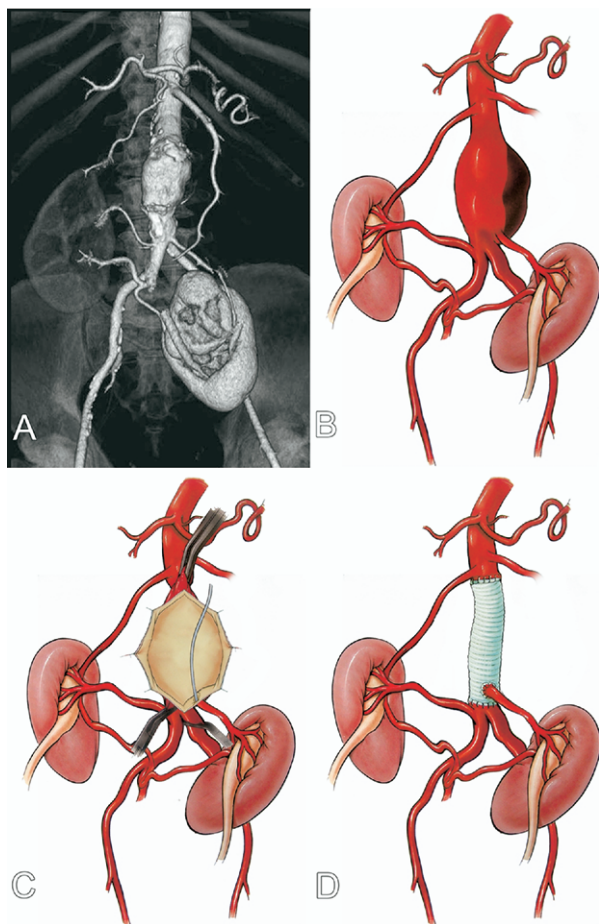


Fig 2. A case of infrarenal aortic aneurysm associated with cPK with a main artery originating from distal aorta and inferior polar artery originating from contralateral hypogastric iliac artery. Preoperative 3D computed tomography scan (A); schematic illustration of stages of the operation: AAA and cPK with a main artery originating from distal aorta and an inferior polar artery originating from right hypogastric iliac artery providing arterial circulation also to the contralateral kidney (B); perfusion of cold crystalloid into the orifices of the cPK artery through an occlusion perfusion balloon catheter (C); end-to-end Dacron bypass with main cPK artery reimplantation (D).

antihypertensive medication was seen in our patients. The patient with the polycystic cPK is alive with a slight worsening of renal function compared with discharge (GFR 39 ml/min compared with 48 ml/min at discharge).

DISCUSSION

Congenital pelvic kidney is the less frequent of the existing six types of renal ectopia (pelvic, lumbar, abdominal, cephalad, thoracic, and crossed), and results from failure of embryological kidney to ascend during fourth to eighth weeks of gestation.¹³

Pelvic kidney is usually small and retains his fetal lobulation. The ureter is usually short and enters the bladder on the same side as the kidney. Fusion abnormalities are rarely

present and, in our knowledge, the association with polycystic dysplasia has never been reported before the present manuscript. The vascular supply is anomalous. One or two renal arteries are common and usually arise from the aortic bifurcation, common iliac artery, and external iliac artery. Rarely, blood supply is guaranteed by feeding arteries originating from inferior mesenteric arteries. Venous and ureteral anomalies are rare. The association of cPK with the previously described anomalies makes an accurate preoperative evaluation mandatory, in order to reduce the risk of iatrogenic lesions during aneurysm isolation and to preserve renal function during aneurysm repair of both cPK and contralateral kidney as previously shown in case 2. Computed tomography and magnetic resonance angiography allow providing information about both aorto-iliac aneurysm anatomy, and cPK feeding arteries and veins. Conventional intra-arterial angiography is an invasive procedure but, recently, as we experienced in our series, allows studying the renal artery anatomy with the highest sensitivity, identifying even small branches or accessory renal arteries that may be misdiagnosed with other techniques. Current technologies in the more recent angio-CT and angio-RM have significantly improved specificity and sensibility of these methods. Duplex ultrasonography provide hemodynamic information such as alterations of peak systolic velocity that can occur in cases of severe kinking or dislocation of cPK renal arteries in cases of huge associated aorto-iliac aneurysms. Retrograde pyelography may provide further information about the path of the ureters and preoperative placement of ureteral catheter can enable the identification of an anomalous ureter.

Because of the presence of cPK, the renal function may be affected by intraoperative renal ischemia after aortic aneurysm repair and several methods of renal protection to prevent renal ischemic injury have been previously reported. These methods of renal preservation, also used during abdominal aneurysmectomy in renal transplant patients, can be divided in four essential groups: pharmacological systemic renal protection, use of a temporary or permanent shunt, in situ perfusion with hypothermic or with pump oxygenator and double proximal clamping technique.¹⁴ The expansion of plasmatic volume with preoperative hydration and the intraoperative administration of furosemide and mannitol¹⁵ or furosemide and dopamine (from 3 to 8 mg/Kg/min in continuous infusion), to obtain a forced diuresis before cross-clamping, reduce the risk of kidney loss, or acute tubular necrosis. These methods are safe and effective and can represent a good choice if associated to a fast aortic reconstruction. The use of a temporary bypass with a Gott shunt¹⁶⁻¹⁸ or a temporary or permanent ipsilateral axillofemoral graft¹⁹⁻²¹ may be effective in preserving a physiological continuous renal perfusion; disadvantages include the possibility of embolization, hemorrhage, and intimal dissection using a Gott shunt; and higher invasivity and groin complications using axillofemoral bypass. To preserve renal function during supra-renal clamping for a longer time (60 to 90 min), it is possible to use selective infusions of bolus of cold (4°C) lactate Ring-

er's solution that can be repeated every 20 minutes or continuously infused in complicated reconstructions.²²⁻²⁶ Another technical approach to this pathology is the renal perfusion using an extracorporeal oxygenation.²⁷ This method offers a reliable protection but requires a perfusionist, a pump-oxygenator, and a groin incision for retrograde cannulation of the femoral artery and vein. Finally, the double proximal clamping technique, described by Lacombe,²⁸ offers a potential sufficient protection for medium time (less than 60 min) using backflow by lumbar, inferior mesenteric, and iliac arteries during completion of proximal aortic anastomosis. The absence of a valid collateral circulation and the presence of a short proximal neck limit the use of this alternative.

In our short series, we experienced the perioperative expansion of plasmatic volume with intraoperative forced diuresis associated with hypothermic selective perfusion of the main cPK arteries with the modalities we used during thoracoabdominal aortic aneurysm repair as effective and safe adjuncts of renal protection.

The occurrence of some amount of preoperative cPK function impairment in case of associated aorto-iliac aneurysms has never been specifically investigated, but it is plausible that the presence of an aorto-iliac aneurysm could affect the function of an associated cPK. Different mechanisms may plausibly be involved in renal function impairment, mainly related to malperfusion of cPK such as aneurysm distal embolization, kinking of the renal artery, hemodynamic effects of abnormal pulsatility, and in the case of large lesions, due to depression of the renal parenchyma, renal veins, and the ureter and/or ureteropelvic junction.²⁹ We unexpectedly experienced a cPK function impairment in three of our patients and, if larger series and specific investigations would be able to confirm this trend, interesting adjunctive issues could be considered in the therapeutic paradigm to indicate aneurysm repair in patients with associated cPK.

CONCLUSIONS

Aorto-iliac aneurysm repair in presence of CPK requires careful preoperative planning and accurate image studies including computed tomography or magnetic resonance angiography, and, in selected case, renal arteries arteriography and duplex ultrasonography, for a complete knowledge of cPK vascular anatomy. In some cases, also renal scintigraphy may be required to study and assess the amount of functioning renal parenchyma.

To preserve renal function during open repair, various methods have been previously reported, but no procedure has been clearly considered the therapeutic gold standard. In our series, the forced intraoperative diuresis with mannitol and furosemide and the main pelvic kidney arteries selective perfusion with hypothermic lactate Ringer solution, associated to a fast surgical technique, have proven to be safe and effective.

In our short series, we reported preoperative cPK function impairment in three out of four cases. Many mechanisms could be hypothesized to justify these findings in the

absence of detectable cPK renal arteries stenosis, but to clarify the roles of the several factors involved, larger series should be analyzed and specific investigations should be performed. In our series, following aorto-iliac aneurysm repair, we observed no worsening of the renal function; besides, we reported a persistent improvement of the renal function in two cases.

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